Ruptured inflammatory abdominal aortic aneurysm: Insights in clinical management and outcome

Andrew L. Tambyraja, MRCSEd, John A. Murie, MD, FRCSG, and **Roderick T. A. Chalmers, MD, FRCSEd,** *Edinburgh, Scotland*

Background: **Ruptured inflammatory abdominal aortic aneurysm (AAA) is relatively rare, and little has been written on the outcome of operative treatment.**
Methods: **Patients undergoing attempted repair of ruptured inflammatory AAA between 1995 and 2001 were included in a retrospective case-cohort study. Demographic, clinical, and operative factors were analyzed, together with in-hospital morbidity, in-hospital mortality, and duration of postoperative hospital stay.**
Results: **Of 297 patients who underwent attempted operative repair of ruptured AAA, 24 (8%) had an inflammatory aneurysm. Twenty-two patients were men, and two were women; median age was 69 years (range, 51-85 years). Operative findings revealed a contained hematoma in 16 patients (70%), free rupture in 3 patients (13%), aortocaval fistula in 4 patients (17%), and aortoenteric fistula in 1 patient (4%). Of 273 noninflammatory ruptured AAAs, only 2 AAA (1%) were associated with primary aortic fistula. Ten patients (42%) with inflammatory AAA died in hospital, compared with 117 of 273 patients (43%) without inflammation. Median postoperative stay was 10 days (range, 0-35 days). Of the 14 patients with inflammatory lesions who survived, 11 had postoperative complications; 4 patients had acute renal failure, three of whom required temporary renal replacement therapy.**
Conclusions: **Ruptured inflammatory AAA is associated with a higher incidence of aortic fistula than is ruptured noninflammatory AAA. Repair of ruptured inflammatory AAA is not associated with increased operative mortality compared with repair of ruptured noninflammatory AAA. (J Vasc Surg 2004;39:400-3.)**

The concept of inflammatory abdominal aortic aneurysm (AAA) was first described by Walker et al[1] to encompass the findings of a thick, white, shiny aneurysm wall and dense fibrosis involving adjacent structures. Reported incidence ranges between 3% and 10%, with a marked male preponderance.[2] Inflammatory AAA presents a surgical challenge because of the difficulty in dissection and control of the proximal and distal aorta.[3] Nevertheless, open repair of intact inflammatory AAA is associated with a similar operative mortality and long-term survival as open repair of noninflammatory AAA.[4-6]

Inflammatory AAA is at lower risk for rupture compared with noninflammatory AAA. Thus it is not surprising that individual surgeons gain little experience in management of ruptured inflammatory lesions.[2,7] We describe the management, clinical outcome, and long-term survival in patients admitted to a large vascular surgery unit with ruptured inflammatory AAA.

From the Department of Vascular Surgery, Royal Infirmary of Edinburgh.
Competition of interest: none.
Presented at the Annual Meeting of the Association of Surgeons of Great Britain & Ireland, Manchester, England, 2003. Published as abstract in Br J Surg 2003;90(suppl 1):119.
Reprint requests: Mr A. L. Tambyraja, Department of Vascular Surgery, Royal Infirmary of Edinburgh, 51 Little France Crescent, Edinburgh EH16 4SA, Scotland (e-mail: andrew.tambyraja@ed.ac.uk).
0741-5214/$30.00

doi:10.1016/j.jvs.2003.07.029

METHODS

All patients admitted to the Edinburgh Vascular Surgical Service for repair of AAA over 7 years from January 1995 to December 2001 were identified from a prospective database and included in a retrospective case-cohort study. The database, together with hospital records, provided demographic details and clinical and operative information for all patients undergoing attempted repair of ruptured AAA. Ruptured aneurysm was defined as presence of retroperitoneal or intraperitoneal blood in the absence of any other identifiable cause for hematoma other than an aneurysm or acute development of an aortic fistula.[8] Inflammatory AAA was diagnosed on the basis of operative appearance. An inflammatory AAA was characterized by the presence of a thickened aneurysm wall, perianeurysm fibrosis, and adhesions to adjacent structures.[2] Histologic confirmation was not routinely obtained. In-hospital morbidity and mortality, length of hospital stay, and postoperative survival were assessed. Survival data were obtained from hospital and general practice records.

Statistical analysis was performed with SPSS for Windows release 11.0.0 (SPSS, Chicago, Ill). Differences between groups were determined with the χ^2 or Fisher exact test for categoric variables and the Mann-Whitney U test for nonparametric continuous variables; $P \leq .05$ was considered significant. Kaplan-Meier life tables were used to calculate survival.

Table I. Presenting symptoms in 24 patients with ruptured inflammatory abdominal aortic aneurysm

Symptom	*No. of patients*
Abdominal/back pain	14
Collapse	8
Hematemesis	1
Hematuria/oliguria/renal failure	1

RESULTS

Patients and demographic data. Five hundred patients underwent attempted repair of nonruptured AAA over the 7-year study. Of these, 54 patients (11%; 43 men and 11 women) had evidence of inflammatory aneurysm. Another 297 patients underwent attempted repair of ruptured AAA during the study, of which 24 aneurysms (8%) were found to be inflammatory.

Patients with ruptured inflammatory AAA included 22 men and 2 women, with median age 69 years (range, 51-85 years). In 3 patients AAA had been diagnosed before presentation, and in 1 of these patients inflammatory AAA was diagnosed before operation. Of these 3 patients one had been considered unsuitable for elective open repair, because of prohibitive ischemic heart disease; 1 male patient had a small aneurysm, with anteroposterior diameter 42 mm on ultrasound scans; and 1 had been deemed unsuitable for open repair, because of the inflammatory process during a previous laparotomy at another unit.

Preoperative findings. Six of the 24 patients were transferred from other hospitals, with a diagnosis of ruptured AAA. Presenting symptoms are described in Table I. In 12 of the 24 patients systolic blood pressure was <90 mm Hg before operation. Preoperatively, 4 patients underwent computed tomography and 3 underwent ultrasound scanning. Computed tomography scans in 2 patients demonstrated features consistent with inflammatory AAA. One patient, with an aortocaval fistula, had established renal failure before surgery. For the group as a whole, preoperative mean serum creatinine concentration was 124 μm/L (range, 68-642 μm/L).

Operative findings. At operation, 16 patients had evidence of a contained retroperitoneal hematoma and 3 had free intraperitoneal blood. Another 4 patients had evidence of aortocaval fistula, and one had a primary aortoduodenal fistula. Of the 273 patients who underwent attempted repair of ruptured noninflammatory AAA over the study period, only 2 patients had a primary aortic fistula, both aortocaval ($P < .0001$). In the group with inflammatory AAA, 20 patients had an infrarenal AAA, 2 of juxtarenal origin, and 2 had evidence of suprarenal extension. In 13 of the 24 patients the aneurysm extended to the iliac artery. No aneurysm was mycotic, as confirmed with subsequent routine microbiologic examination of mural thrombus.

Table II. Postoperative morbidity after successful repair of ruptured inflammatory abdominal aortic aneurysm in 14 patients

Morbidity	*No. of patients*
Renal failure	4
Left ventricular failure	3
Deep vein thrombosis	2
Myocardial Infarction	1
Perforated duodenal ulcer	1
Respiratory tract infection	1
Ileus	1
Limb ischemia	1

Operative management. Operative technique in all operations was through a transperitoneal approach, with minimal dissection of adherent structures from the aneurysm. Two patients with suprarenal lesions and 1 patient with an aortoduodenal fistula died intraoperatively, and no prosthesis was inserted. Of the 21 remaining patients, infrarenal aortic clamping was performed in 19 and in 2 patients a balloon occlusion catheter was required for proximal control. Standard operative practice entailed avoidance of supraceliac clamping whenever possible. Eleven patients received a bifurcated graft, and 10 received a straight graft. All aortocaval fistulas were closed off from within the AAA sac with polypropylene sutures. In 7 patients careful dissection and mobilization of the left renal vein was required, and in 1 of these patients the vein was ligated. No patient underwent ureterolysis, and there were no iatrogenic enteric injuries.

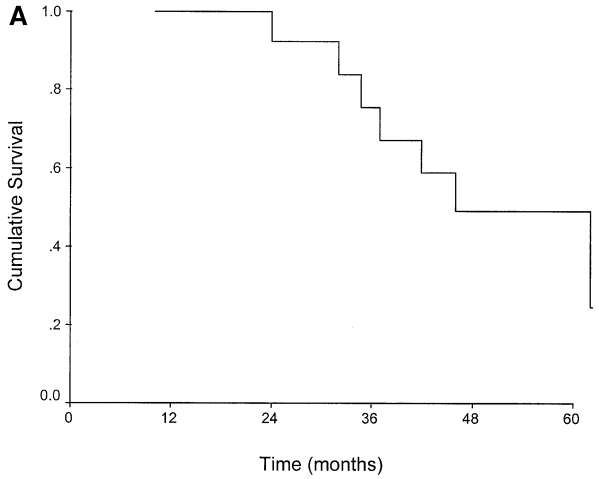
Postoperative outcome. In-hospital mortality in the inflammatory AAA series was 10 of 24 patients (42%). Seven deaths were intraoperative or within 24 hours of surgery, and three deaths occurred later. Of 273 patients admitted with ruptured noninflammatory AAA during the study period, 117 patients (43%) died in hospital ($P = .910$), 69 intraoperatively or within 24 hours of surgery ($P = .495$).

Of the 14 surviving patients in the inflammatory AAA group, 11 had one or more postoperative complications (Table II). Three patients required further surgery, one to treat postoperative bleeding, one for femoral embolectomy, and one for repair of a perforated duodenal ulcer. Of 156 surviving patients in the noninflammatory AAA group, 125 developed one or more postoperative complications ($P = .889$).

Four patients developed acute renal failure after repair of ruptured inflammatory AAA, and in 3 of these temporary renal replacement therapy was required. No patient required permanent renal support. Of 4 patients who underwent repair of aortocaval fistulas, postoperative lower limb deep vein thrombosis developed in 2 patients. None died.

Median postoperative hospital stay for survivors of ruptured inflammatory AAA was 12 days (range, 9-35 days), compared with 13 days (range, 8-129 days) for survivors of ruptured noninflammatory lesions ($P = .422$)

A



B

Time (months)	0	12	24	36	48
Number at risk (Cumulative survival)	14	13 (100%)	12 (92%)	9 (76%)	4 (49%)

Postoperative survival after successful repair of ruptured inflammatory abdominal aortic aneurysm in 14 patients.

Long-term survival. Median survival of the 14 patients discharged from the hospital was 42 months (range, 10-62 months; Fig). In 1 patient late renal failure developed, but this was not secondary to obstructive nephropathy. No patient was discharged with steroid therapy, and none have had postoperative graft complications.

DISCUSSION

To our knowledge, there are no other contemporary series specifically describing management and outcome of ruptured inflammatory AAA. The current retrospective case-cohort study describes a single-center experience in the management of such aneurysms.

It is recognized that the concept of inflammatory aneurysm does not entail a distinct clinicopathologic entity, but one end of a spectrum of chronic inflammatory changes seen in degenerative aneurysms.[9] However, all patients in the current series had the defining triad of thickened aneurysm wall, extensive perianeurysm fibrosis, and dense adhesions to adjacent organs, as described by Rasmussen and Hallett.[2]

Risk for rupture of inflammatory AAA is generally thought to be lower than that of noninflammatory AAA.[2,7] Nevertheless, natural progression to enlargement and eventual rupture persists.[1,2,7,9,10] Case-cohort studies have reported the observed incidence of rupture to be between 3% and 4% in inflammatory AAA, compared with 17% to 20% in noninflammatory AAA, but these figures stem from relatively small series.[9,10] The present data show that the relative proportions of inflammatory and noninflammatory AAA that present with rupture do not exhibit such disparity; the true rate of rupture of inflammatory AAA may be closer to that of noninflammatory lesions than previously assumed.

Previous reports suggested that the combination of lower rupture risk and higher perioperative mortality after elective repair renders correct management of asymptomatic inflammatory AAA unclear.[10] While it is impossible to determine the true rate of incidence and rupture of inflammatory lesions without population-based screening and prospective follow-up, the incidence of ruptured inflammatory AAA in the current series does not support such a conservative stance.

The incidence of obstructive uropathy in inflammatory AAA is generally reported to be approximately 20%.[4,9] It is interesting that in the current series no patient had evidence of significant renal failure that could be attributed to obstructive nephropathy. Nitecki et al,[5] in a case-cohort study of 29 intact inflammatory AAAs, reported no significant differences in preoperative serum creatinine concentration or incidence of chronic renal failure compared with patients with noninflammatory AAA. However, patients with inflammatory aneurysms were at increased risk for postoperative renal failure, and 11% required renal replacement therapy. In the present series no patient had clinical evidence of obstructive nephropathy; and only one had renal failure, secondary to an aortocaval fistula. Despite this, 5 of 24 patients had postoperative renal failure and required renal replacement therapy. However, it is recognized that the etiology of postoperative nephropathy is quite different after repair of ruptured AAAs compared with intact lesions.

Aortovenous fistula complicating rupture of an AAA has an incidence of approximately 2% to 4%.[11,12] Death after operative repair of spontaneous aortocaval fistula has been reported at 21% to 55%.[13] From the present data, only 1% of noninflammatory AAAs were associated with aortocaval fistula, as opposed to 17% of inflammatory aneurysms. Of interest, the only primary aortoenteric fistula in the current series was in a patient with a ruptured inflammatory AAA. Calligaro et al,[11] in a review of aortovenous fistula associated with ruptured AAA, cited four reports of aortovenous fistula associated with inflammatory lesions, and thought this reflected an increased predisposition for posterior rather than anterior rupture, as a result of anterior fibrosis and thickening. Together with recognition that periaortic fibrosis causes adherence to posterior veins in 63% to 70% of inflammatory AAAs, an increased incidence of aortovenous fistula is unsurprising.[4]

None of the 4 patients with aortocaval fistulas in the present series died in hospital, although repair was associated with postoperative deep venous thrombosis in 2 patients, despite subcutaneous unfractionated heparin thromboprophylaxis. Postoperative deep venous thrombosis is a noteworthy complication after repair of aortocaval fistula. Measures to minimize risk may include therapeutic anticoagulation postoperatively, followed by a period of oral anticoagulation after discharge. It seems that heightened

awareness of the possibility of aortovenous fistula should exist in management of ruptured inflammatory AAA. Simple closure of the fistula, together with insertion of an aortic prosthesis, can be performed with acceptable morbidity and mortality.

It is accepted that elective or urgent open repair of intact inflammatory AAA can be performed with mortality that approaches that of noninflammatory AAA repair.[3,4,6] Outcome after repair of ruptured inflammatory lesions is less well-recorded. Lindblad et al[10] reported an operative mortality rate or 75% in 4 patients with ruptured inflammatory AAA, compared with 44% in 16 patients with ruptured noninflammatory AAA, but it is impossible to derive a meaningful conclusion from such a small series. The present larger series exhibited a 42% operative in-hospital mortality rate, similar to the operative in-hospital mortality rate of 43% for ruptured noninflammatory AAA during the study period.

Few analogous series describe the outcomes of endovascular repair of inflammatory AAA. Hinchliffe et al,[14] in a series of 14 patients with intact inflammatory AAA, reported a 14% mortality rate, although a potential role in patients with previous failed open repair is proposed. Furthermore, potential for endovascular stent grafting to exacerbate perianeurysm inflammatory fibrosis has been described by Vallabhaneni et al.[15] Although it is proposed that endovascular techniques offer an alternative to open repair of stable ruptured AAA, indications remain unclear in both noninflammatory and inflammatory lesions.[16]

In conclusion, despite the operative challenge associated with repair of ruptured inflammatory lesions, the inflammatory process itself has no appreciable immediate effect in terms of outcome.

We thank Mr A. McL. Jenkins, Professor A. W. Bradbury, Mr J. Sathianathan, and Mr D. G. Kitts for allowing us to report on their patients.

REFERENCES

1. Walker DI, Bloor K, Williams G, Gillie I. Inflammatory aneurysms of the abdominal aorta. Br J Surg 1972;59:609-14.
2. Rasmussen TE, Hallett JW Jr. Inflammatory abdominal aortic aneurysms: a clinical review with new perspectives in pathogenesis. Ann Surg 1997;225:155-64.
3. Sultan S, Duffy S, Madhavan P, Colgan MP, Moore D, Shanik G. Fifteen year experience of transperitoneal management of abdominal aortic aneurysms. Eur J Vasc Endovasc Surg 1999;18:510-4.
4. Pennell RC, Hollier LH, Lie JT, Bernatz PE, Joyce JW, Pairolero PC, et al. Inflammatory abdominal aortic aneurysms: a thirty-year review. J Vasc Surg 1985;2:859-69.
5. Nitecki SS, Hallett JW Jr, Stanson AW, Ilstrup DM, Bower TC, Cherry KJ, et al. Inflammatory abdominal aortic aneurysms: a case-control study. J Vasc Surg 1996;23:860-9.
6. Crawford JL, Stowe CL, Safi HL, Hallman CH, Crawford ES. Inflammatory aneurysms of the aorta. J Vasc Surg 1985;2:113-24.
7. Goldstone J, Malone JM, Moore WS. Inflammatory aneurysms of the abdominal aorta. Surgery 1978;83:425-30.
8. Bradbury AW, Makhdoomi KR, Adam DJ, Murie JA, Jenkins AMcL, Ruckley CV. Twelve-year experience of the management of ruptured abdominal aortic aneurysm. Br J Surg 1997;84:1705-7.
9. Sterpetti AV, Hunter WJ, Feldhaus RJ, Chasan P, McNamara M, Cisternino S, et al. Inflammatory aneurysms of the abdominal aorta: Incidence, pathologic and etiologic considerations. J Vasc Surg 1989; 9:643-50.
10. Lindblad B, Almgren B, Bergqvist D, Eriksson I, Forsberg O, Glimaker H, et al. Abdominal aortic aneurysm with perianeurysmal fibrosis: Experience from 11 Swedish vascular centres. J Vasc Surg 1991;13: 231-9.
11. Calligaro KD, Savarese RP, DeLaurentis DA. Unusual aspects of aortovenous fistulas associated with ruptured abdominal aortic aneurysms. J Vasc Surg 1990;12:586-90.
12. Brewster DC, Cambria RP, Moncure AC, Darling RC, LaMuraglia GM, Geller SC, et al. Aorto-caval and iliac arteriovenous fistulas: recognition and treatment. J Vasc Surg 1991;13:253-65.
13. Alexander JJ, Imbembo AL. Aorto-vena cava fistula. Surgery 1989;105: 1-12.
14. Hinchliffe RJ, Macierewicz JA, Hopkinson BR. Endovascular repair of inflammatory abdominal aortic aneurysms. J Endovasc Ther 2002;9: 277-81.
15. Vallabhaneni SR, McWilliams RG, Anbarasu A, Rowlands PC, Brennan JA, Gould DA, et al. Perianeurysmal fibrosis: a relative contraindication to endovascular repair. Eur J Vasc Endovasc Surg 2001; 22:535-41.
16. Hinchliffe RJ, Braithwaite BD, Hopkinson BR. The endovascular management of ruptured abdominal aortic aneurysm. Eur J Vasc Endovasc Surg 2003;25:191-201.

Submitted Jun 05, 2003; accepted Jul 31, 2003.